www.jmscr.igmpublication.org
Index Copernicus Value: 79.54
ISSN (e)-2347-176x ISSN (p) 2455-0450
crossref DOI: https://dx.doi.org/10.18535/jmscr/v7i6.123

IGM Publication
Journal Of Medical Science And Clinical Research
An Official Publication Of IGM Publication

Case Report

Papillary Cystadenocarcinoma of Submandibular Gland- A Case Report

Authors
Parveen Rana Kundu[1], Amrita Kulhria[2*], Swaran Kaur[3], Neha Salaria[4], Ruchi Agarwal[5], Reena Verma[6]

[1,5]Associate Professor, Department of Pathology, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana
[2]Senior Resident, Department of Pathology, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana
[3]Professor & Head, Department of Pathology, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana
[4]Assistant Professor, Department of ENT, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana
[6]Junior Resident, Department of Pathology, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana.
*Corresponding Author
Amrita Kulhria
Addresses: Department of Pathology, B.P.S. Government Medical College For Women, Khanpur Kalan, Sonipat Haryana. 131305, India

Abstract
Introduction: *Papillary cystadenocarcinoma is an extremely rare malignant neoplasm of major salivary glands. The most common major salivary gland involved is parotid. It is characterised by cysts and papillary endophytic projections.*
Case Report: *We report a case of papillary cystadenocarcinoma arising from the left submandibular gland in a 55-year-old female patient, and discussed the clinical, histopathological and treatment features of this rare entity.*
Conclusion: *Papillary Cystadenocarcinoma is a very rare malignant tumor of the salivary gland. It is to be differentiated from other papillary tumors of salivary gland especially papillary cystadenoma.*
Keywords: *Papillary cystadenocarcinoma, Major salivary gland.*

Introduction

Papillary cystadenocarcinoma is an extremely rare, malignant tumor of major salivary gland comprising 2% of all salivary gland malignancies.[1] It was first defined as distinctive entity by WHO in 1991.[2] It is also known by the terminology such as malignant papillary cystadenoma, low-grade papillary adenocarcinoma or mucus-producing adeno papillary carcinoma.[3] In this article, we report an unusual case of papillary cystadenocarcinoma arising from the left submandibular gland.

Case Report

A 55-year-old female patient presented to ent opd with painless, slow growing mass in the left submandibular region. Computed Tomography revealed a mass lesion in the left submandibular gland. Fine needle aspiration was performed and diagnosis of salivary gland neoplasm with cystic change was made. The patient underwent left submandibular gland excision with adherent soft tissue.

Grossly, received soft tissue mass measuring 7x3.5x2 cm. On serial slicing, a relatively circumscribed variegated growth measuring 4.3x3x1.8 cm was revealed (Figure 1). It was partly solid and partly cystic. Papillary excrescences were noted. Seven lymph nodes were isolated from adherent tissue.

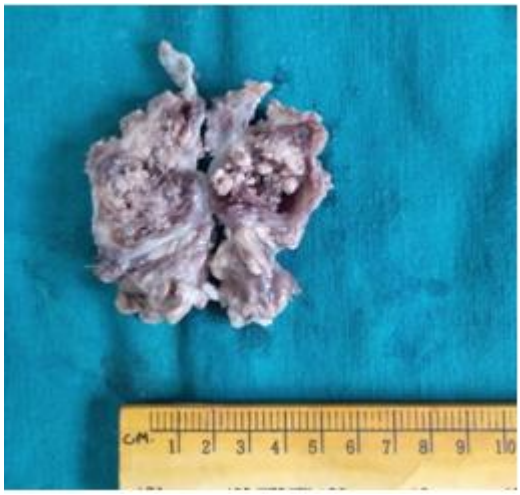

Figure (1): Well circumscribed partly solid and partly cystic papillary growth in salivary gland parenchyma

Microscopy revealed minimally invasive cystic tumor. The cystic spaces showed variable sized papillary structure lined by cuboidal to columnar cells with clear to vacuolated cytoplasm (Figure 2). The tumor cells showed mild to moderate degree of pleomorphism along with focal areas of invasion. Foci of calcification were also seen at places. The tumors cells were positive for cytokeratin (Figure 3). All lymph nodes were free from tumor. Histolopathological diagnosis of Papillary Cystadenocarcinoma (low grade) was made.

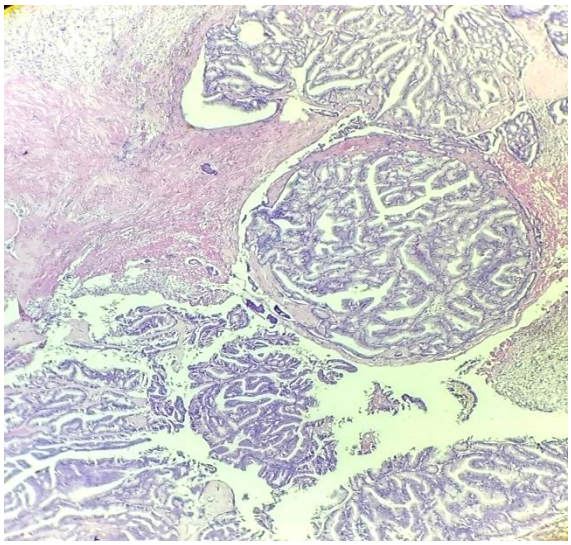

Figure (2): Microphotograph showing cystic cavities filled with papillary projections (H&E; 4X)

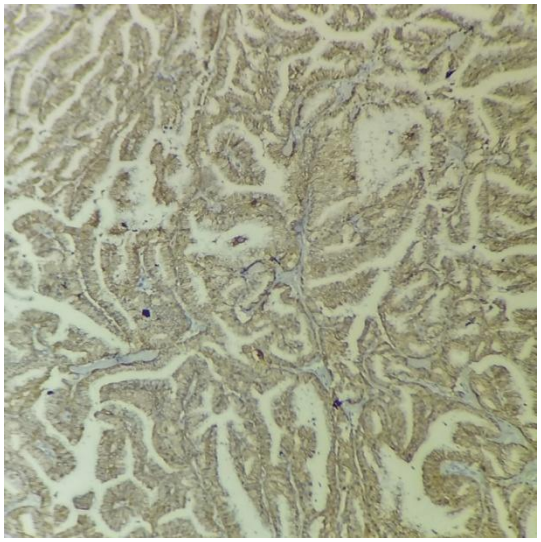

Figure (3) Cells positive for CK (H&E; 10X)

Discussion

Salivary gland papillary cystadenocarcinoma represents a distinct group of malignancy in major and minor salivary glands. They usually have an indolent biologic behaviour but there have been several reports which indicates aggressive behaviour of the tumor.[4] The most common sites are major salivary gland and mainly parotid gland.[5] According to study by Foss et al, there was no gender preference and the mean age of presentation was 57 years.[6] Both high and low-grade cystadenocarcinomas have been documented in the literature.[4,7,8] Low grade tumor are characterised by relatively uniform nuclei and rare mitoses.[9] High grade tumours

show nuclear pseudo stratification, high nuclear cytoplasmic ratio, numerous mitotic figures and the presence of areas of necrosis. Lymphovascular, perineural invasion and meatastasis to lymph nodes indicates towards high grade of tumor.[10] The management is complete surgical excisions and radiotherapy in high grade tumor.

The differential diagnosis of papillary cystadenocarcinoma includes cystadenoma, papillary variant of acinic cell carcinoma, metastatic papillary carcinoma, polymorphous low-grade adenocarcinoma, low grade mucoepidermoid carcinoma.[3,4,11] Papillary cystadenoma is characterised by monomorphic or mild atypical tumor cells and abscence of infiltration in surrounding areas. Infitration of tumor was present in our case which ruled out this entity. Papillary variant of acinic cell carcinoma shows characterstic acinar differentiation.[9] Metastatic papillary carcinoma thyroid was excluded in virtue of normal radiological reports. Low grade mucoepidermoid carcinoma may be cystic but is characterized by the presence of squamoid cells, mucus cells and cells of intermediate type.[12] Absence of characterstic pattern of polymorphous low-grade adenocarcinoma ruled out this also. So, all above differentials were excluded before making definitive diagnosis of papillary cystadenocarcinoma of submandibular gland.

Conclusion

Papillary Cystadenocarcinoma is a very rare malignant tumor of the salivary gland. It is to be differentiated from other papillary tumors of salivary gland especially papillary cystadenoma. These tumours have been reported to have good prognosis. Follow up is necessary because of their propensity for local recurrence and metastases even after many years.

Financial support and sponsorship: Nil.

Conflicts of interest: There are no conflicts of interest

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